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**Acute neonatal respiratory distress caused by a lingual thyroid: the role of nasendoscopy and medical treatment**

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Note

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## **Abstract**

Background: Lingual thyroid is a known cause of oropharyngeal obstruction in the neonate, and can be asymptomatic or present as stridor, dysphonia, dysphagia and dyspnoea with faltering growth. The therapeutic options include surgical resection.

Case report: A 6-day-old female neonate, born at 36 weeks gestation presented with stridulous breathing and poor feeding. Although initially thought to be laryngomalacia, nasendoscopy revealed a lingual thyroid. The baby had deranged thyroid function detected on neonatal screening but this result was not available until a later date. Despite being symptomatic, the patient was managed medically and thyroxine therapy was associated with resolution of the respiratory symptoms.

Conclusion: Nasendoscopy provides valuable information about an ectopic thyroid gland, and thyroid replacement therapy may help to suppress the size of the ectopic gland and ultimately prevent an unnecessary surgical procedure.

## **Keywords**

Lingual Thyroid, Airway Obstruction, Endoscopes, Thyroxine, Infant, Newborn

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## **Introduction**

Around 80% of babies with congenital hypothyroidism (CHT) will have an underlying dysgenesis where the gland has failed to develop and / or descend normally. Thyroid dysgenesis is sporadic, with a female predominance<sup>1</sup>. A lingual thyroid is one particular subgroup of dysgenesis and occurs in approximately 1:100,000 children. In this instance the thyroid gland has failed to descend to the usual pretracheal region<sup>2</sup>. Babies may be asymptomatic but may also present with oropharyngeal obstruction including stridor, dysphonia, dysphagia and dyspnoea with faltering growth. Thyroid ectopia may be confirmed by ultrasound and/or thyroid isotope scan<sup>3</sup>, and up to 70% of patients with lingual thyroid are hypothyroid<sup>4</sup>. An ectopic thyroid gland may therefore be suspected in a baby with respiratory compromise who also has a positive screening test for CHT. Management options for babies in severe respiratory distress include surgical resection of the ectopic thyroid gland.

## **Case report**

A female infant presented on several occasions to the Accident and Emergency department and the postnatal ward in the first ten days of life because of stridulous breathing, worse when supine and during feeding. She was born at 36 weeks gestation via normal vaginal delivery and was her healthy parents' first child. Her birth weight was 2.43kg (25<sup>th</sup> centile). She was thought to have laryngomalacia and sent home on each occasion until day eleven when she was admitted and reviewed by the tertiary respiratory team. By this stage she weighed 2.25kg and had noisy breathing with significant associated respiratory effort.

The baby was seen by the ENT team and underwent a nasendoscopy. This revealed a mass on the tongue that was approximately 2 x 2 cm in size and covered in mucosa. The baby had

undergone neonatal screening for conditions that included CHT on day five of postnatal life. However, there was insufficient bloodspot sample to conduct the necessary triplicate TSH analysis. Analysis of a single blood spot had revealed an elevated value of 24.3mU/l ( $\geq 20$  is screen positive), but a repeat sample was requested rather than an 'abnormal' result reported and hence the clinical team were not aware of this background. In the interim the baby had undergone magnetic resonance imaging (MRI) of her neck following the ENT assessment. The MRI confirmed a mass within her tongue (Figure 1), in keeping with an ectopic lingual thyroid gland. This diagnosis was confirmed by technetium isotope scan (Figure 2) and formal thyroid function tests on serum conducted on day 21 of life indicated CHT before the repeat blood spot sample had been collected. The TSH was elevated at 53.4mU/l with a Free T4 of 16.2pmol/l, which is at the lower end of the newborn reference range<sup>5</sup>.

The baby was commenced on oral thyroxine 25 micrograms daily (10mcg/kg). The aim was to administer a substantial dose despite there being good evidence of endogenous thyroid gland production. The objective was to reduce TSH levels to less than 1mU/l.

The stridor and her bottle and breast-feeding improved in the subsequent days, and the baby was discharged with scheduled out-patient follow-up. On review three months after starting thyroxine, the patient remained asymptomatic and continued to grow well with no feeding problems. Her TSH 13 days post intervention with thyroxine was 0.3mU/l.

## **Discussion**

Lingual thyroid is a well-recognised cause of respiratory distress and our case highlights the importance of medical as well as surgical intervention in this scenario. Patient age and symptoms are key considerations when deciding on an optimal therapeutic strategy<sup>2</sup>.

Management may require surgical removal by direct excision and ablation with radioisotopes<sup>2</sup>, although neither is ideal in the neonate. Surgical resection does have high success rates, but there are significant risks associated with anaesthesia in addition to concerns about inflammation, scarring and incomplete resection. The use of radioactive isotopes in children may be a less invasive option, but the risks associated with radiation exposure need to be considered carefully<sup>6</sup>.

Dutta *et al.*<sup>7</sup> reported three cases of older children with dysphagia and odynophagia. All three children were diagnosed by ultrasound and technetium scan. They responded well to treatment with levothyroxine alone, which resolved the associated compressive symptoms by reducing the size of the ectopic thyroid tissue. TSH will promote the growth of thyroid tissue, and removing this stimulus with thyroxine replacement will result in a reduction in gland size<sup>8</sup>. Our experience suggests that the response can be relatively rapid – over a matter of days rather than weeks, although a general increase in airway circumference may be another key factor that contributed to the improvement. There are very few case reports where medical treatment was the only therapy for lingual thyroid associated with compressive symptoms<sup>7,9</sup>.

The most important diagnostic tool has been suggested to be the thyroid technetium scan, which enables detection of ectopic thyroid tissue<sup>2,3</sup>. Ultrasonography, computed tomography (CT) and MRI are helpful in defining the extent and location of the ectopic thyroid gland. Nasendoscopy, as used in the case described, may be used with or without general anaesthesia and provides views of the nasal fosse, choanae, pharynx and larynx, even in the neonate. In this case, the nasendoscopy was carried out successfully without the use of general anaesthesia.

There will always be occasions when repeat samples are requested as part of a screening programme, and it was unfortunate that this baby not only had an ectopic thyroid gland but was one of those where the TSH sample was insufficient. It is important for laboratories to adhere to a predetermined set of standards, appreciating there will be a tension between the desire to provide rapid feedback and the desire to provide reliable information. Closer links between the laboratory and the clinical team may have resulted in a repeat sample being obtained at an earlier stage. These oversights are an inevitable part of clinical practice, and highlights the fact that the newborn screening programme should be recognised for what it is – a screening, not a diagnostic programme. Therefore, clinicians should maintain a clinical awareness of congenital hypothyroidism and not assume that the screening programme will have detected all cases.

In summary, nasendoscopy can give valuable information in babies when there is little external evidence of thyroid ectopia, and prompt intervention with thyroxine in the baby with thyroid ectopia and associated respiratory compromise, can result in a rapid response and minimise the need for surgical intervention. We would recommend a substantial dose of thyroxine so that TSH suppression occurs relatively quickly, removing the stimulus for further gland growth.

## Figures



Figure 1: MRI demonstrating mass (11 x 8 x 8 mm) at the base of the tongue

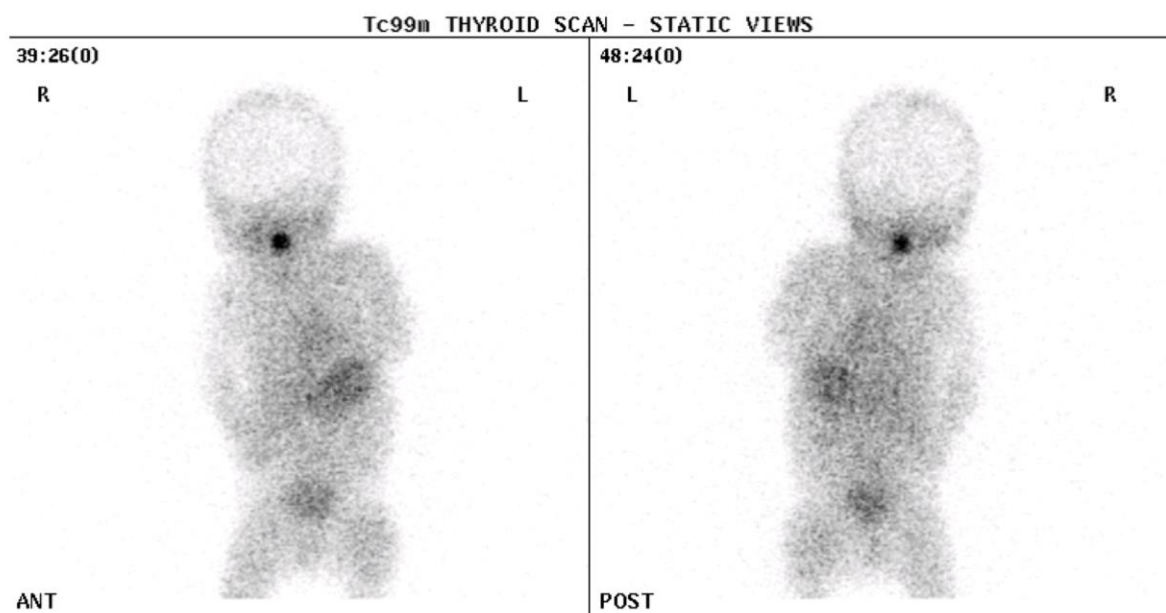


Figure 2: Technetium isotope scan highlighting ectopic thyroid tissue



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## Summary

- Lingual thyroid is a rare embryological anomaly, where the thyroid gland has failed to descend to the usual pretracheal region.
- Ectopic lingual thyroid should be considered in the differential diagnosis of infants presenting with evidence of oropharyngeal obstruction and poor feeding.
- Nasendoscopy can give valuable information about an ectopic thyroid gland.
- Thyroid replacement therapy may help to suppress the size of the ectopic thyroid gland and prevent an unnecessary surgical procedure.
- Surgery should only be considered if adequate thyroid replacement therapy fails to resolve obstructive symptoms.